

Review Article

Neonatal tooth with bilateral cleft lip and palate: A case report with review of literature

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ABSTRACT

Natal and neonatal teeth are present in the oral cavity at birth and during the first 30 days of life, respectively. Management of such teeth is a treatment challenge. The incidence of such natal and neonatal teeth is reported to be 1 in every 1125-30,000 births, respectively. Cleft lip and palate (CLP) is a common dentofacial congenital anomaly of the oral cavity and may be present unilaterally or bilaterally. The occurrence of association of such teeth with unilateral and bilateral CLP cases is reported to be 2% and 10%, respectively. The presence of natal/neonatal teeth in the cleft area complicates the child's health only in few cases. There is a paucity of information in the literature regarding the simultaneous occurrence of neonatal teeth in bilateral CLP cases. The present paper reported the management of a neonatal tooth present in association with complete bilateral CLP in a 12-day-old male child with review of literature.

Key words: Cleft lip, Cleft lip and palate, Cleft palate, Neonatal tooth

Eruption of a tooth is a milestone from both functional and psychological aspects of a child's life. Chronology of tooth eruption is subject to variations depending on hereditary, endocrine and environmental factors. Sometimes the first teeth may be present at birth or erupt during the first month of life which is termed as predeciduous teeth, dentition praecox, fetal teeth, or congenital teeth [1]. Massler and Savara in 1950 classified these teeth by taking only the time of eruption as reference. They defined natal teeth as those present in the oral cavity at birth and neonatal teeth as those erupting during the first 30 days of life. This classification has been widely used till date [2]. Some investigators have also reported the association of natal and neonatal teeth with various syndromes as well such as Hallerman-Streiff syndrome, Ellis-Van Creveld syndrome, craniofacial dysostosis, steatocystoma multiplex, pachyonychia congenita, and sotos syndrome [3-6].

Cleft lip and palate (CLP) is one of the most common congenital anomalies in the orofacial region of a newborn which may be unilateral or bilateral. Various dental anomalies are particularly common in patients with CLP such as supernumerary teeth, natal and neonatal teeth, ectopic eruption, agenesis, cone-shaped teeth, hypoplasia, hypocalcification, and intranasal teeth [7-11]. A strong relation has been found with the occurrence of anomalies in size, shape, number, structure, and eruption of teeth in the cleft area [10,12]. Although a high prevalence of natal/

neonatal teeth in the cleft area is observed, only a small number of epidemiological reports have been published [13,14].

Comparatively very few case reports have been presented with the occurrence of natal and neonatal teeth in association with CLP cases. The present paper depicts a case report describing the presence of neonatal tooth in a patient with complete bilateral CLP along with the review of literature.

CASE REPORT

A 12-day-old male child with CLP was referred from the Department of Pediatrics to the Department of Pediatric and Preventive Dentistry of our college with the presence of a solitary tooth like structure in the upper jaw since 7 days after birth. The tooth was causing feeding difficulty for the child and he often cried refusing the intake of milk. The parents, however, were worried that the tooth appeared as bad omen. The myth was immediately cleared, and the parents were educated regarding the occurrence and management of the neonatal tooth. The medical history was non-contributory. The male infant was born at term through cesarean section. The mother was not exposed to any agents known to be mutagenic or teratogenic during her pregnancy. Infant was healthy with no signs of any systemic disorder.

On clinical examination, a single tooth like structure was present in the left side of premaxillary segment having grade II

mobility. The tooth was irregular, larger in size (macrodontia), and yellowish in color with black deposits around the cervical region (Fig. 1). Since the tooth erupted 7 days after birth a diagnosis of neonatal tooth was established. Medical examination revealed no systemic abnormalities. Extraction of the tooth was performed under topical anesthesia. This extracted neonatal tooth was having only crown portion without any root structure (Fig. 2). Follow-up after 7 days of extraction was uneventful.

DISCUSSION

The presence of a tooth in the oral cavity during or shortly after birth is referred to as natal and neonatal tooth, respectively. Natal teeth develop in the intrauterine period and are present in the infant's mouth during birth, while neonatal teeth erupt after birth within 30 days of life. The incidence of natal and neonatal teeth varies from 1 in every 1125-30,000 births with a higher occurrence seen with natal teeth than neonatal teeth [15]. 85% of the natal/neonatal teeth are mandibular incisors and are found to be more common in females [7,16]. Table 1 depicts the prevalence of natal and neonatal teeth as reported in the literature by various authors [3].

Approximately, 90% of the reported natal or neonatal teeth are the primary teeth, while the rest are supernumeraries [7]. The etiology of natal or neonatal teeth is still controversial as many factors have been proposed to explain the possible etiology. Several reasons have been put forth such as heredity, endocrine disturbances, infections such as congenital syphilis and nutritional deficiencies like hypovitaminosis, etc. [14,17-19]. Sometimes fever, exanthemata during pregnancy can accelerate the eruption process and it is also dependent on osteoblastic activity within the area of the tooth germ [15]. Superficial position of the tooth germ may also be the cause in some cases [4,7]. Possible complications that arise from the presence of natal teeth include discomfort during suckling, aspiration of the teeth into his/her airway and lungs, interference in retaining the pre-surgical orthodontic

appliances in cleft babies, abrasions of the maternal nipple and ulceration to the ventral surface of the child's tongue with refusal to feed (Riga-Fede disease) [20-22].



Figure 1: Presence of a single tooth like structure on the left side of the pre-maxillary segment which was seen to be yellowish in color with black deposits around the cervical region

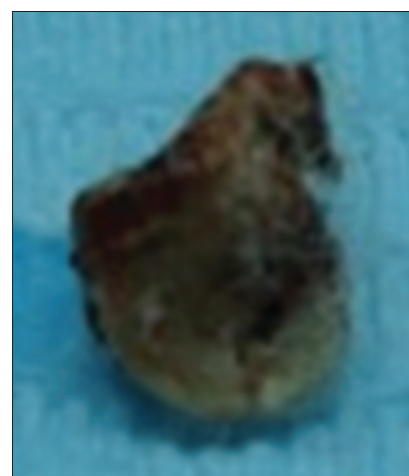


Figure 2: The extracted neonatal tooth having only crown portion without any root structure

Table 1: Natal and neonatal teeth with cleft lip and palate reported in the literature with their associated problems and management

Authors	Prevalence %	Sample size	Associated problems	Management
Ameida et al. 1996	2.02-10.6	1019	Aspiration of mobile natal/neonatal teeth	Extraction of all teeth
Mandana et al. 2005	Not mentioned	-	Teeth interfered with the fabrication and application of the NAM appliance	Extraction of teeth
Iwamoto et al. 2009	9.1	55	Aspiration of mobile natal/neonatal teeth and risk of traumatic ulceration on the lip	Extraction of teeth
Manjushree et al. 2013	1.98-2.8	151	Aspiration of mobile natal/neonatal teeth and difficulty in feeding	Extraction of teeth
Nagaveni et al. 2015	6.6	15	Aspiration of mobile natal/neonatal teeth and during pre-surgical orthopedic therapy, the neonatal teeth may impede the fit of the intraoral plate and require adjustments	Extraction of teeth
Yilmaz et al. 2016	7	65	Aspiration of mobile natal/neonatal teeth and during pre-surgical orthopedic therapy, the neonatal teeth may impede the fit of the intraoral plate and require adjustments	Extraction of all teeth performed before the impression stage to prevent any aspirations

NAM: Nasoalveolar molding

However, authors have suggested that there is no relationship between injury to mother's nipple and the presence of the natal teeth since the tongue is interposed between these teeth and the nipple during breastfeeding [21]. These babies are also brought to dental clinic due to the myth of bad omen or devil's incarnation or to know whether the tooth is part of the normal dentition or is supernumerary [22]. In our case, the child was brought to the pediatric department as he was experiencing difficulty in feeding along with the potential risk of inhaling the tooth. The parents also believed in the myth of bad omen due to the presence of the neonatal tooth.

CLP is one of the most common congenital anomalies in the orofacial region of a newborn which may be unilateral or bilateral. Worldwide incidence of CLP is 1:600 live births [23]. The term CLP describes the potential complexities of the deformity which may involve nose, lips, alveolus, and palate. It affects breathing, appearance, dentition, facial growth, speech, and hearing leading to psychosocial implications [24].

The prevalence of natal or neonatal teeth was found to be significantly higher in patients with CLP [20]. The high prevalence could be due to dental disturbances related to the alveolar clefts and superficial localization of teeth in this region [5]. No significant difference has been stated between genders in some studies [25], while Ronk mentioned that these teeth are more frequent in females [26]. Similarly, Kates et al. found the presence of natal teeth to be double in females as compared to males [27]. On the other hand, some researchers argued that this situation occurred more frequently in males or that no gender differences are evident [3].

Cabete et al. carried out the evaluation of primary dentition in children who had CLP with and without natal/neonatal teeth. Missing primary lateral incisor in the cleft region was the most prevalent anomaly in children with natal/neonatal teeth. It was found to be 52% in unilateral complete cleft cases and 62% in bilateral complete cleft cases. In their study, they also correlated the occurrence of supernumeraries, missing lateral incisors and normal dentition in children with unilateral and bilateral clefts with natal/neonatal teeth, compared to those without natal/neonatal teeth. They found no significant difference in their occurrences [15].

Moreover, several authors also suggest that in patients with CLP, the natal/neonatal teeth occur more often in maxilla and in primary lateral incisor region near the lateral side of the cleft [6]. De Almeida and Gomide reported that the prevalence of natal and neonatal teeth in unilateral CLP was 2.02% and in bilateral CLP was 10.06% [7]. Iwamoto et al. have reported the prevalence of natal and neonatal teeth to be 5.1% and 30% in unilateral and bilateral CLP, respectively. According to them in cleft patients, the space for the tooth germ to exist is insufficient due to poor development of maxillary ridge owing to the cleft; as a result, the tooth germ might be located in a more superficial area of the residual ridge on lateral side of the cleft [28].

The treatment of patients with CLP starts soon after birth. After adequate clinical evaluation, the intraoral impression is routinely taken for preparing the feeding plate. Consequently,

the presence of natal/neonatal teeth can impede the impression making. Hence, first, the natal/neonatal teeth have to be treated before the impression stage to prevent any aspiration. The duration of the pre-surgical orthopedic therapy is approximately 3-4 months. If teeth emerge after birth, the intraoral plate may not fit and modifications of the plate may be required. In some cases, the therapy has to stop and postponed until the teeth have been extracted [25]. Thus, timely management of natal/neonatal teeth is highly necessary for the proper management of clefts.

In the management of neonatal teeth, sometimes a dental radiograph may be helpful in differentiating whether the tooth is a prematurely erupted deciduous tooth or a large soft tissue enlargement [29,30]. In the view of several authors, all neonatal teeth should be extracted [31]. Yilmaz et al. showed that all the neonatal teeth were located at the cleft-side and consequently inside the borders of the intraoral molding plate. Therefore, they suggested that neonatal tooth extraction should be performed routinely in all cases with CLP who will undergo pre-surgical orthopedic treatment. The extraction is performed not earlier than 10 days after birth. During the first 10 days after delivery, the bacterial flora may be ineffective in Vitamin K production, which plays a major role in prothrombin synthesis. The risk of hemorrhage due to hypoprothrombinemia is present. To avoid any complications, prothrombin blood tests are also reviewed before any intervention [25]. Basavanthappa et al. advised Vitamin K administration if extraction due to mobility has to be performed before the age of 10 days. Pre-surgical orthopedic treatment is started after approximately 1-week of the healing period [30].

At times, the healing period can be prolonged and during the follow-up period only horizontal lip bands may be used. In some cases, neonatal teeth emerge during the 1st month of life after the start of pre-surgical orthopedic treatment or the systemic well-being of the child does not enable the extraction. In such cases, modifying the border of the intraoral plate until extraction and during the soft tissue healing period to enable fit of the plate may be recommended [25].

In the present case, a neonatal tooth was present on the left side in the premaxilla having grade II mobility. The tooth was causing difficulty in feeding and also there was potential risk of aspiration and swallowing. The extraction of the tooth was planned and carried out under topical anesthesia. Since the child was 12-day-old, extraction was carried out on the same day. Followed by this, a feeding appliance was planned and fabricated to improve feeding and facilitate proper respiration.

CONCLUSION

As a clinician, one should be aware of all the factors related to conditions such as the presence of natal or neonatal teeth and CLP, which will help in correct diagnosis and treatment planning. It will also allow us to consider the adverse effects these teeth have for infants as well as their parents thereby helping us in its adequate management.

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